# 6

# Other Consequences of Clefts

Speech-language pathologists (SLPs) treating children with clefts need to be aware of three areas of concern, in addition to those discussed in previous chapters: (1) dental and orthodontic issues; (2) hearing problems; and (3) social, educational, emotional, and psychological issues. Each of these has an impact on the well-being of the child and family and affects how well the youngster will be doing when he or she reaches young adulthood.

## DENTAL AND ORTHODONTIC ISSUES

Children with clefts are likely to have missing teeth, malpositioned and malaligned teeth, and malocclusions. All these conditions are treatable, but the original condition and sometimes the treatment itself can cause articulation problems, if only temporarily. The SLP who gets a new referral of a patient with a cleft needs some basic knowledge of the following: (1) dental development in children; (2) the terminology that dental specialists use with regard to the primary dentition and the permanent dentition; and (3) how likely it is that a given dental abnormality, malocclusion, or treatment device may contribute to a speech problem.

### Normal Development of Dentition

The *primary dentition (milk teeth, deciduous dentition)* consists of 20 teeth. The lower central incisors erupt first, and then the remaining teeth erupt according to the sequence shown in Figure 6-1. Most children have all their primary teeth in place by the age of 2.5 years. By the age of 6 or 7 years, the mixed dentition phase begins, during which the primary teeth are gradually replaced by permanent teeth.

There are usually 32 teeth in the *permanent dentition*; this number depends on whether the third molars or *wisdom teeth* (the most posterior teeth on each side of each dental arch) erupt (Fig. 6-2). In general, eruption of the mandibular permanent teeth is slightly ahead of eruption of the maxillary teeth.

### Occlusion

The main points for describing how the maxillary and mandibular dental arches fit together are the first molars. That is, the orthodontist categorizes the occlusion

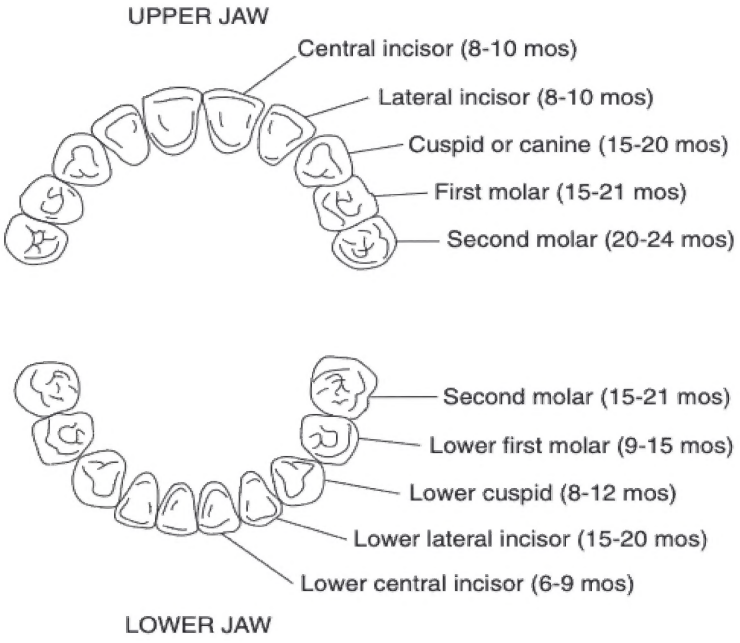
UPPER JAW
Central incisor (8-10 mos)
Lateral incisor (8-10 mos)
Cuspid or canine (15-20 mos)
First molar (15-21 mos)
Second molar (20-24 mos)
Second molar (15-21 mos)
Lower first molar (9-15 mos)
Lower cuspid (8-12 mos)
Lower lateral incisor (15-20 mos)
Lower central incisor (6-9 mos)
LOWER JAW

**FIGURE 6-1** The primary dentition and the timing of normal tooth eruption. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

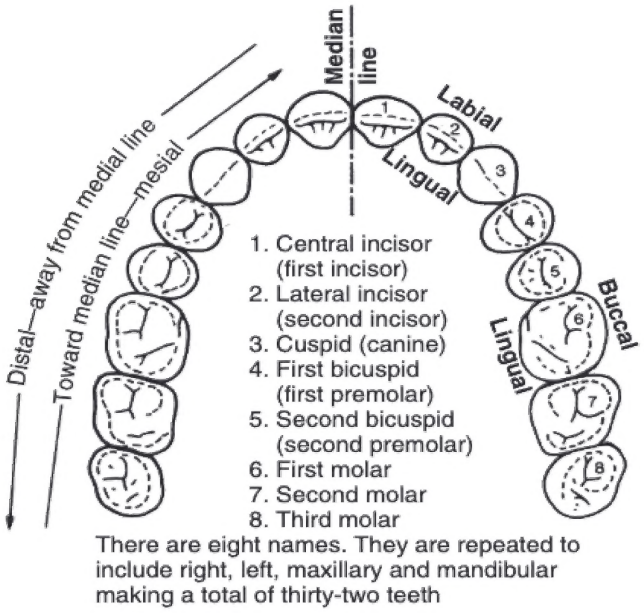
Median line
Labial
Lingual
Distal—away from medial line
Toward median line—mesial
Buccal
Lingual
1. Central incisor (first incisor)
2. Lateral incisor (second incisor)
3. Cuspid (canine)
4. First bicuspid (first premolar)
5. Second bicuspid (second premolar)
6. First molar
7. Second molar
8. Third molar
There are eight names. They are repeated to include right, left, maxillary and mandibular making a total of thirty-two teeth

**FIGURE 6-2** The permanent dentition in an adult dental arch and terms used for orientation (distal, medial, labial, buccal). (From Wheeler RC: *Textbook of dental anatomy and physiology*. Philadelphia: Saunders, 1940, p 7.)

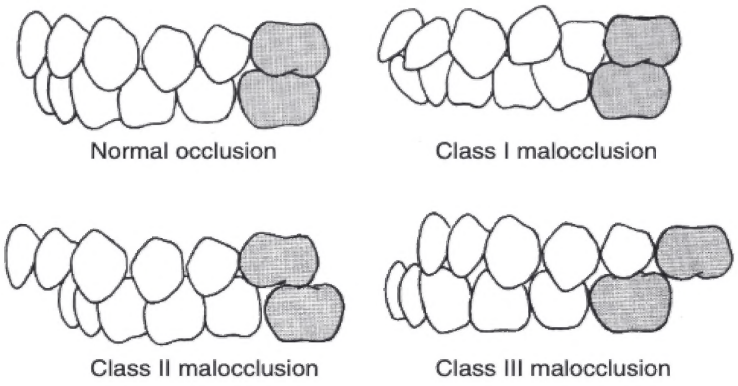


**FIGURE 6-3** Normal occlusion and malocclusion classes as specified by angle. (From Proffit WR: *Contemporary orthodontics* (3rd ed). St. Louis: Mosby, 2000, p 4.)

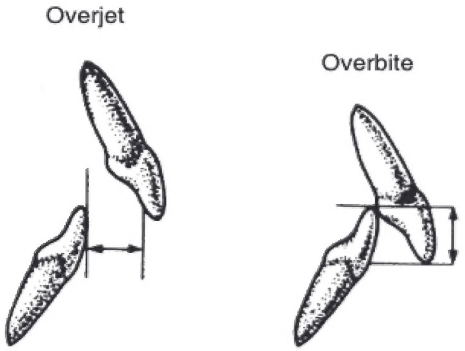


**FIGURE 6-4** Relationship between the upper and lower incisors in overjet and overbite. (From Hall DJ, Warren DW: Orthodontic problems in children. In Bluestone CD, Stool SE [eds]: *Pediatric otolaryngology,* vol 2. Philadelphia: Saunders, 1983, p 966.)

according to how the upper and lower first molars are positioned in relation to each other. The maxillary arch is normally slightly wider than the mandibular arch and to some extent fits over it (Fig. 6-3). The maxillary teeth are expected to have about a half-tooth overlap with their counterparts in the mandibular arch, and the molar occlusion is such that the inner cusps (the cusps nearer the tongue) of the upper teeth fit into the central grooves between the cusps in the lower teeth. In normal occlusion, there is a slight overjet and overbite of the anterior maxillary teeth in relationship to the anterior mandibular teeth, as shown in Figure 6-4. Table 6-1 is a list of additional terms used in the description of occlusion (Figs. 6-5 through 6-10).

## Open Bites

Anterior open bites (see Fig. 6-9) often occur in young children and are usually not related to clefts. The most common causes are nasopharyngeal airway obstruction (e.g., due to hypertrophic adenoids) and thumb sucking. Both of these are examples of "function changing form," rather than form causing problems in function. When the nasopharyngeal airway is obstructed, the child becomes an obligate oronasal breather. A chronic mouth-open posture in a child can lead to

**TABLE 6-1**

*Common Terms Used to Describe Dental and Occlusal Anomalies*

| TERM | DESCRIPTION |
|---|---|
| Crossbite | Maxillary teeth inside, or *lingual* to, mandibular teeth |
| Anterior crossbite (see Fig. 6-5) | Maxillary incisors behind mandibular incisors; may describe single teeth or all incisors |
| Canine crossbite | Crossbite of the canine teeth |
| Lateral crossbite, also called buccal or lingual crossbite (see Fig. 6-6) | Crossbite of the teeth in the lateral segment or segment nearest the cheek |
| Neutrocclusion, class I molar relationship (see Fig. 6-3) | The *sagittal* (front-to-back) relationship of the jaws is normal but there may be malalignment of individual teeth or a discrepancy in jaw position in the transverse or horizontal plane |
| Class II molar relationship (see Figs. 6-3 and 6-7) | Maxillary teeth more anterior to the mandibular teeth than normal with a larger overjet; often accompanied by excessive vertical overlap of the maxillary incisors over the mandibular incisors; also called *closed bite, deep bite*, or *overbite* |
| Class III molar relationship (see Figs. 6-3 and 6-8) | Maxillary molars posterior to mandibular molars; usually accompanied by anterior *crossbite* or *underbite*, although sometimes the anterior mandibular teeth are tipped backward, or *retroclined*, so that they fit behind the anterior maxillary teeth* |
| Anterior open bite (see Fig. 6-9) | Open space between the anterior maxillary and mandibular teeth; often the result of finger habit or habitual open-mouth breathing |
| Posterior open bite (see Fig. 6-10) | Open space between the maxillary and mandibular teeth (molars or molars plus bicuspids) |

From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* (4th ed). St. Louis: Mosby, 2010.
*As one may suspect, nondentists often confuse the terms that should be used to describe only the molar relationship (class I, class II, class III) with the terms used to describe the relationship between the anterior maxillary and mandibular teeth (overbite, overjet, underbite, crossbite).

overeruption of the posterior teeth, eventually propping the mouth open. Thus, even if the nasopharyngeal airway obstruction is resolved by natural adenoid involution, the anterior teeth may be prevented from meeting. Another consequence of a chronic mouth-open posture is a physical change in the palatal vault: Because apposition of the tongue surface against the palatal vault is not normal (due to the fact that the child keeps the mouth open to breathe), inward or medial collapse of the lateral palatal segments, hypertrophy or overdevelopment of soft tissue, or both, can occur (Fig. 6-11).

There is also posterior open bite, in which the posterior teeth of the maxillary and mandibular arch do not meet. In theory, this could lead to lateral air emission in articulation. Unilateral posterior open bite is common in hemifacial microsomia until all aspects of orthodontic treatment are completed.

**FIGURE 6-5** Patient with an anterior crossbite. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

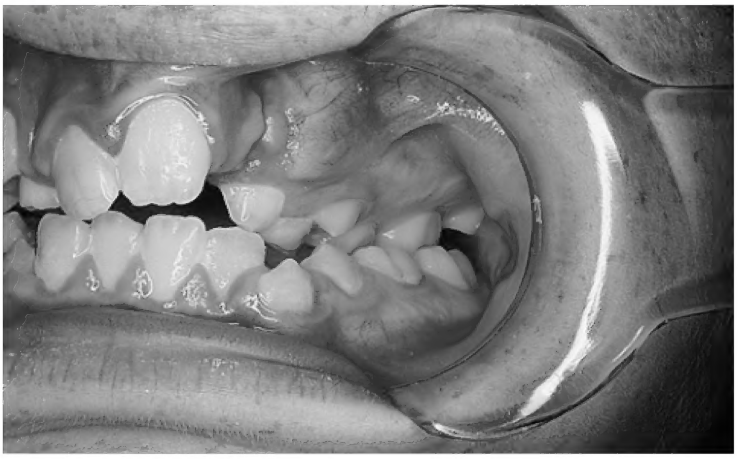

**FIGURE 6-6** Patient with a canine and buccal crossbite. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

**FIGURE 6-7** Patient with a class II malocclusion. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

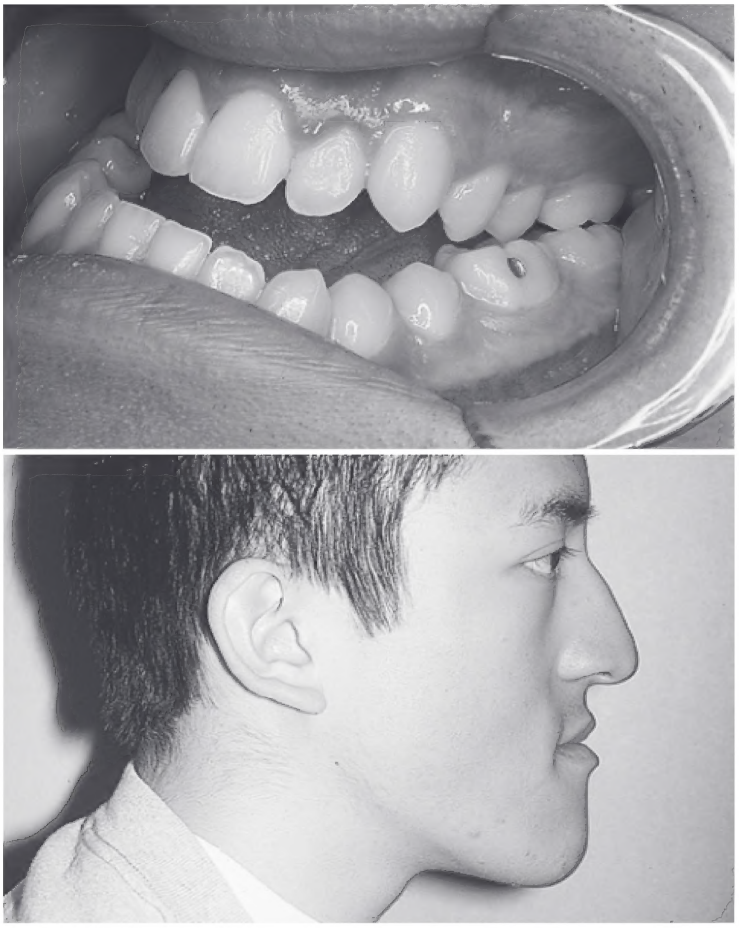

**FIGURE 6-8** Intraoral and profile views of a patient with a class III malocclusion. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

"Mouth breather" is a commonly used term; however, unless nasal airflow is documented by aerodynamic studies to be totally absent, it cannot be assumed all the airflow is going in and out of the mouth simply because the child breathes with the mouth open. Thus, the preferred term is "oronasal breather."

This alteration in shape is often seen in children who are obligate oronasal breathers. Clinicians typically apply the term "high-arched palate" and unwisely assume that any speech problems they hear are attributable to this shape.

The palate is described as "high-arched" because of the central groove that is seen between the medially displaced or hypertrophic palatal shelves, but this should not be considered a diagnostic term unless there is radiographic proof that the palatal plane is higher than it should be. Furthermore, there are no data indicating a relationship between this palatal shape and any given speech problem. If there were such a relationship, the most likely problem would be articulation issues resulting from inadequate "operating space" for the tongue, not hypernasality or nasal air loss.

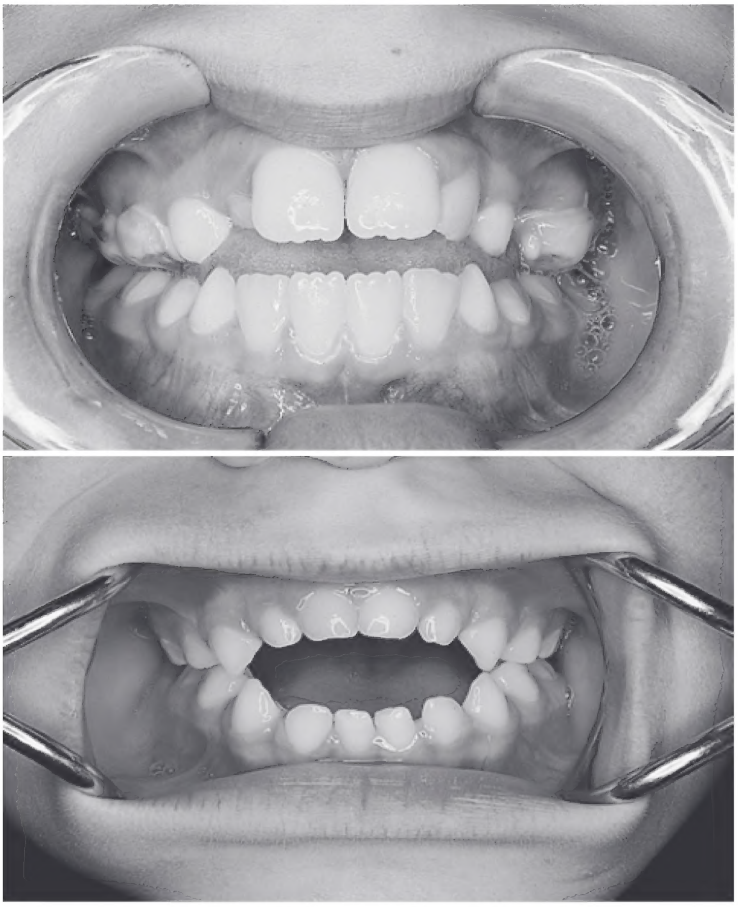

**FIGURE 6-9** Two patients with anterior open bites, one more severe than the other. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

**FIGURE 6-10** Patient with a posterior open bite. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

**FIGURE 6-11** The intraoral view that often leads clinicians to use the term *"high-arched palate"*. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

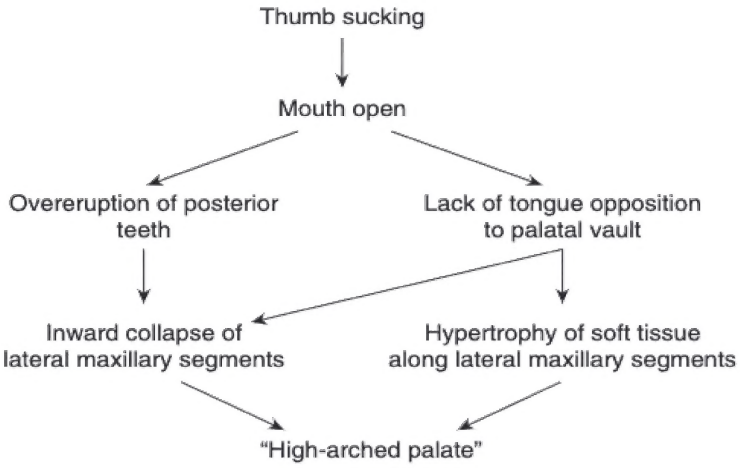
Thumb sucking
Mouth open
Overeruption of posterior teeth
Lack of tongue opposition to palatal vault
Inward collapse of lateral maxillary segments
Hypertrophy of soft tissue along lateral maxillary segments
"High-arched palate"

**FIGURE 6-12** Sequence of events cascading from thumb sucking. This sequence leads to the subjective impression of a high-arched palate, often mistakenly assumed to be a cause of speech problems. (Modified from Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

A similar sequence of events can be initiated by thumb sucking, which orthodontists tend to label "a finger habit." Figure 6-12 is a diagram of the consequences.

## Dentition and Occlusion in Cleft Palate

### A Special Treatment Issue for Infants With Clefts: Presurgical Orthopedics

For many decades, orthodontists and surgeons have tried various ways of manipulating the palatal segments to make both lip closure and palatal closure easier. Many different devices have been proposed, from relatively simple lip taping (Fig. 6-13) to the use of both passive and active intraoral plates (Figs. 6-14 and 6-15).

**FIGURE 6-13** Taping to bring the premaxilla into a more advantageous position for lip repair in two babies with bilateral clefts. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

**FIGURE 6-14** An intraoral plate for partially obturating a bilateral cleft in an infant and for preventing inward collapse of the lateral segments. (From Rutrick R, Black PW, Jurkiewicz MJ: Bilateral cleft lip and palate: presurgical treatment. *Ann Plast Surg* 12:105-117, 1984.)

Passive plates made to prevent inward collapse of the lateral palatal segments may also assist feeding, depending on the configuration of the cleft. Active plates consist of two palatal segments joined by a screw mechanism. The screw can be turned to move the segments outward very gradually, thereby correcting arch collapse. See Figures 6-14 and 6-15.

Presurgical orthopedics (also called infant orthopedics or "IO") has been highly controversial since at least the early 1960s. Both passive and active plates have been shown to have no long-term effect on the position of the palatal segments and may in fact prevent the natural addition of tissue to the inner surfaces of the palatal shelves in the first few months of life. (Please see Chapter 2 for

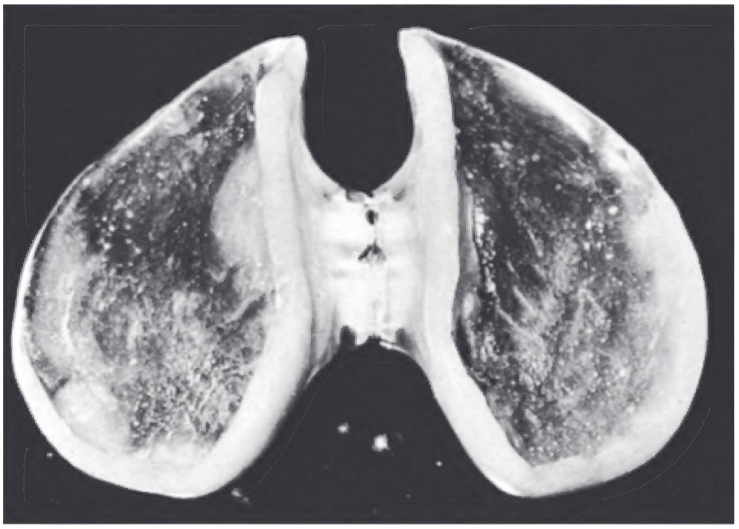

**FIGURE 6-15** An expansion plate fabricated for a baby with a bilateral cleft in whom the palatal shelves had collapsed toward the midline. (From Rutrick R, Black PW, Jurkiewicz MJ: Bilateral cleft lip and palate: presurgical treatment. *Ann Plast Surg* 112:105-117, 1984.)

comments on "feeding" plates.) Peterson-Falzone (2012) summarized the results from 24 papers published since 1999 on passive and active infant plates, including the Dutch cleft studies and reports on *nasoalveolar molding* (NAM; discussed later). The predominance of the evidence at that time (2012) was that infant orthopedics produced no long-term positive results or improvement in treatment outcomes.

Lip taping is less controversial, less expensive, and less demanding on parents, and it seems to cause less stress on infants. However, it does require several presurgical visits to the treating team to monitor both the skin beneath the tape and the position of the lip segments. Interestingly, Mother Nature rules: Even when the lip segments have been brought into good alignment for surgery, the tape is not removed until the baby reaches the operating room because, once the tape is off, the segments begin to move back to their original position. This is particularly true in bilateral clefts.

A popular but still controversial form of presurgical orthopedics called *nasoalveolar molding* (NAM) is used by many teams. This treatment consists of an intranasal stent attached to an intraoral plate (Matsuo et al., 1989; Matsuo and Hirose, 1991; Grayson et al., 1999). At night while the baby sleeps, the stent is inserted into the nostril on the cleft side for the first 3 to 12 months of life, during which time it reshapes, expands, and repositions the external nasal structures. At the same time, the intraoral plate repositions and realigns the palatal segments.

Clinicians who use NAM believe that it makes the initial surgical closures of the lip, nose, and palate easier because it puts the divided segments into closer proximity to each other and minimizes the scar tissue associated with surgery; others have found that its benefits are short-lived (e.g., the nasal tissue returns to its collapsed position), and the treatment is expensive in dollars, child

discomfort, and associated family stress. It should be noted that the treatment can be carried out only when the family can make weekly visits to the team during the months of treatment. (However, some families very much appreciate the opportunity to be active participants in their baby's care.) Although the treatment was developed in the late 1980s, there are still no long-term data proving that the final outcome in children who had the NAM treatment is measurably better than the outcome in those who did not receive it. To our knowledge, no NAM studies have been conducted with an untreated control group. Currently there are many treatment centers throughout the United States that use NAM, particularly for bilateral clefts of the lip and palate.

### Dental Anomalies

Dental development may be delayed in individuals with clefts; this pertains particularly to the teeth close to the alveolar cleft. In addition, there can be (1) missing teeth (meaning they never develop because the tooth buds are absent), (2) duplicated or supernumerary teeth, and (3) malpositioned teeth. In clefts that affect the alveolar ridge, it is easy to understand that the presence of the cleft may contribute to absence of tooth buds, but even children with clefts of the palate alone have been found to have a significant occurrence of missing teeth (Ross, 1975). Duplicated teeth are regarded as the result of the cleft's "splitting" a tooth bud, resulting in two teeth instead of one. Children with clefts also have a high incidence of missing teeth *outside* the cleft area (Lekkas et al., 2000). All these dental anomalies are treatable and need not affect speech on a long-term basis. Orthodontic treatment planning generally begins when the permanent dentition erupts: malpositioned teeth can be moved orthodontically, and missing teeth can be replaced.

Replacement of missing teeth may be accomplished with removable plates, fixed bridges, or implants. Implants are much more expensive and thus more difficult to convince third-party payers to cover. Extra teeth are not routinely removed but are instead are used to take up the space left by missing teeth (e.g., a missing lateral incisor or canine).

In young children with clefts, the spaces created by missing teeth or teeth that are late in erupting may contribute to oral distortion of complex consonants such as sibilants and affricates, but the effect is usually temporary. Speech therapy to correct tongue placement on these consonants in the youngster is not recommended because nature or the dental specialist will fill the space. If deviant tongue placement in articulation persists after the teeth are in place, then therapy should be instituted. The same could be said for youngsters who are undergoing orthodontic therapy for malocclusions: wait until treatment is complete, and then reassess speech.

Sadly, dental health of children in the United States is too often *not* a matter of concern to parents, and children frequently present in cleft palate or craniofacial clinics with decayed teeth that have rarely been brushed. The treating SLP in the home community can help by encouraging the family to follow recommendations for regular oral hygiene (meaning teeth cleaning by a dental hygienist at least every 6 months) and checkups by the dentist.

## Malocclusions

For nondentists, it is confusing that orthodontists assess two aspects of occlusion: the relationship between the jaws and the relationship between the teeth in the jaws. The former is called the *skeletal* relationship and the latter is the *dental* relationship. For speech-language pathologists, the difference may seem obscure and trivial, but dental problems are more easily treatable (by orthodontics) than are skeletal problems (often treatable only by jaw surgery) and thus are less likely to have long-term effects on speech.

The most common malocclusions in individuals with clefts are various forms of *crossbite* (maxillary teeth inside the mandibular teeth). Patients with unilateral cleft lip and palate often have a crossbite on the cleft side (Figs. 6-16 and 6-17). Another term for this condition is *arch collapse*. Even if the patient has only a unilateral cleft lip and palate, a "bilateral" crossbite may be present if many teeth are missing in the maxilla, as seen in Figure 6-17.

Patients with bilateral clefts may present with a bilateral crossbite or arch collapse, a condition that is particularly likely if a poor, aggressive palate repair caused extensive scar tissue in the palate and effectively "locked out" a protrusive premaxilla. (In the patient pictured in Figure 6-18, orthodontics should have been used to move the palatal segments outward and bring the premaxilla into appropriate position.)

*Class II malocclusions* (maxillary teeth more anterior to the mandibular teeth than normal, with a larger than normal overbite) are rare in clefts, except in those more complex conditions such as Pierre Robin sequence, mandibulofacial dysostosis, and Stickler syndrome. An example of a class II malocclusion is seen in Figure 6-7.

In *class III malocclusions* (maxillary first molars posterior to their mandibular counterparts; usually accompanied by an anterior crossbite or *underbite*), the mandible appears prognathic (see Fig. 6-8). Patients with clefts are particularly vulnerable to class III malocclusions for two reasons, one of which is a subject of a long-term controversy. Orthodontists generally agree that, in patients with

**FIGURE 6-16** Patient with a crossbite of the teeth nearest the cleft only, with a normal molar relationship. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

**FIGURE 6-17** This patient had only a left unilateral cleft lip and palate but has a bilateral crossbite, a condition caused in part by multiple missing teeth in the maxillary arch. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

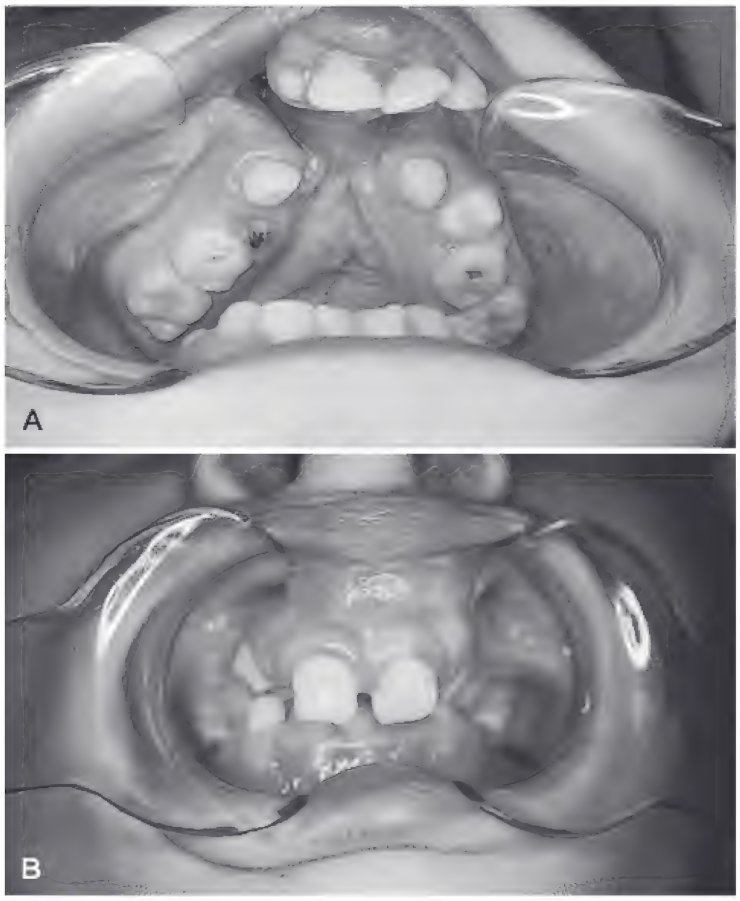


**FIGURE 6-18** Both these patients have a severely protrusive premaxilla, with the lateral segments collapsed behind it. In **A,** note the alveolar defects and the pile-up of scar tissue from the palatal repair in the palatal vault. The alveolar clefts are also unrepaired in **B,** but the segments abut one another. (From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* [4th ed]. St. Louis: Mosby, 2010.)

clefts, maxillae that are restricted in size and retruded in position are the result of both inherent tissue deficiencies and surgery that has further restricted growth, particularly if the surgery was overly aggressive and resulted in extensive scar tissue.

It is generally agreed that aggressive techniques of surgical closure of the lip and palate, such as those that result in a very tight lip, collapsed palatal arches, and a shallow palatal vault with thick scars, lead to maxillary underdevelopment and class III malocclusions. Individuals with this type of unfortunate surgical outcome are likely to become candidates for another type of treatment known as *maxillary advancement surgery*, which is just one category of *orthognathic surgery*. ("Ortho" means "straight" and "gnathic" means jaws.)

Fear of interfering with growth potential of the midface is the main reason for delaying surgical closure of the palate. In several treatment centers, including some in the United States, the palate is left open into early childhood and even school-age years. This treatment approach is known as *primary veloplasty* because the soft palate is closed in infancy or early toddlerhood, while the hard palate is left open. Please see Chapter 3 on physical management.

Good et al. (2007) reported an overall frequency of 20.9% need for maxillary advancement in 207 patients with cleft lip and palate or cleft palate only.

A class III malocclusion that is so severe that the lower teeth are far ahead of the lower teeth can affect speech in several ways: (1) labiodentals are produced in an "inverted" fashion, with the lower teeth contacting the upper lip; (2) sibilants, affricates, and alveolar stops may be distorted because the tongue is so far ahead of the palatal vault that the speaker has difficulty attaining appropriate articulatory placement (in extreme cases, intended tip-alveolars become not just "tip-dentals" but "tip-labials"); (3) bilabials may become quasilabiodentals because the two lips are so mismatched in position. Occasionally these speakers are misdiagnosed as having abnormally large tongues simply because more of the tongue is visible. Remember the tongue is literally rooted in the mandible. Where the mandible resides skeletally, so does the tongue.

## Maxillary Advancement as Treatment for Class III Malocclusions

The maxilla can be advanced either surgically or through distraction osteogenesis. The latter is a combination surgical-orthodontic treatment that accomplishes the goal more slowly than does one-time orthognathic (jaw) surgery. Clinicians who perform either orthognathic surgery or distraction osteogenesis to treat class III malocclusions make very careful measurements of just how much advancement is needed. If these clinicians are very thorough, they will also measure how the maxillary advancement may affect velopharyngeal (VP) closure.

In distraction osteogenesis of the maxilla, a surgeon first separates the maxilla from its attachment to the pterygoid plate and then makes a transverse (horizontal) incision high above the tooth roots to detach the maxilla from the floor of the nose. A mechanical distraction device, which can be either internal (intraoral) (Kumar et al., 2006) or external

(Polley and Figueroa, 1997) is attached to the maxilla and gradually moved forward by turning screws a very small amount each day or twice a day. New bone fills in between the separated bony surfaces (think of the old adage that "nature abhors a vacuum"). This is the osteogenesis part. A single maxillary advancement surgical procedure accomplishes the same goal by moving the maxilla all the way forward to the desired position at surgery and fixed in that position with the use of plates and screws. The orthodontist places a custom-made splint to hold the maxilla in that position until healing is complete. Neither type of maxillary advancement should be undertaken before the mandible has reached full growth (approximately age 17 years for girls, 18 years for boys). After advancement, the maxilla will not grow forward (Huang et al., 2007); if the mandible is still growing, there will be "relapse" into the previously existing malocclusion.

Multiple reports have described the development of incomplete VP closure for speech in patients with clefts who had competent VP mechanisms before maxillary advancement, whether the advancement was accomplished by a surgical procedure alone (i.e., a LeFort I maxillary osteotomy [Chanchareonsook et al., 2006; Haapanen et al., 1997; Kummer et al., 1989; Maegawa et al., 1998; Mason et al., 1980; Okazaki et al. 1993; Watzke et al., 1990]) or by distraction osteogenesis (Chanchareonsook et al., 2006; Guyette et al., 2001). It may seem strange that clinicians have reported a lack of correlation between the amount of linear advancement of the maxilla (as measured radiographically) and the onset of inadequate VP closure. However, as pointed out by Kummer et al. (1989), variables in post-procedure muscle function such as velar stretch and increased inward motion of the lateral pharyngeal walls may compensate for increased depth of the VP port. Figure 6-19 shows a potential change in the dimensions of the VP port (as a result of maxillary advancement) that could conceivably result in the onset of VP inadequacy.

## Summary for Speech-Language Pathologists

SLPs working on cleft palate or craniofacial teams quickly learn that there is often little correlation between the extent of a physical "defect" (variation from the norm) and how well the speaker does in terms of articulation, airflow control, and resonance. The physical situation and the parameters of speech output must be evaluated on an individual basis. Dental and occlusal anomalies are rarely

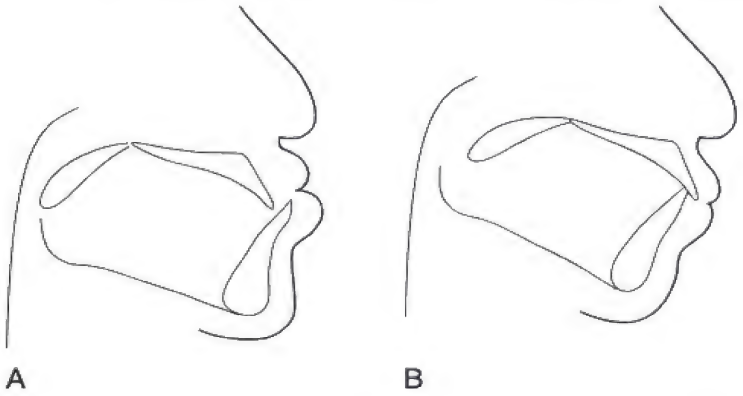


**FIGURE 6-19** **A** and **B,** Potential changes in the dimensions of the velopharyngeal port (as a result of maxillary advancement) that could conceivably result in the onset of velopharyngeal inadequacy.

serious or long-term threats to speech, but the SLP needs to know the terminology and what others have learned from their clinical experience.

## EAR DISEASE AND HEARING LOSS

Children typically have bouts of earaches, signaling the presence of otitis media, and fluctuating hearing levels. Children with clefts are even more vulnerable to otitis media because of the interlink between muscles of the palate and the muscular system that opens the eustachian tube, thus allowing normal aeration of the middle ear. In a child with an overt or submucous cleft palate, that muscular system cannot function properly.

### Physical Basis of the Problem

The muscle that carries the primary responsibility for opening the eustachian tube is the tensor palatini (Hixon et al., 2013). In the presence of a cleft, the tensor does not have its normal points of origin or insertion and thus cannot open the eustachian tube orifice with enough "oomph" (see Chapin and Bluestone, 2009 and Peterson-Falzone et al., 2010, for extensive references on this topic). This means a reduced ability for air to enter the middle ear space. Whatever air is already in the space is absorbed by the mucous lining of the cavity, creating negative air pressure. The negative air pressure in the middle ear cavity retracts the eardrum inward, which is very painful for the child. The negative pressure also draws bacteria up through the tube from the pharynx into the middle ear. Other factors that contribute to poor tubal function include abnormal anatomical positioning of the cartilage of the tube with respect to the tensor muscle (Shibahara and Sando, 1988) and poor quality of the cartilage itself, making the tube "floppy" and more collapsible (Bluestone, 1971; Bluestone et al., 1975; Bluestone et al., 1972; Matsune et al., 1991; Moore et al., 1986). The poor quality of the cartilage helps to explain why patients with adequate VP muscle function after palatal surgery often continue to present with tubal dysfunction and otitis media (da Silva et al., 2010).

The effects of the anatomical and functional deficiencies in the eustachian tube are seen very early in infants with clefts: More than 90% are born with fluid in the middle ear (Paradise et al., 1969; Stool and Randall, 1967). If the otitis media goes untreated or repeatedly returns, complications can include infections of the tube, tympanic membrane perforations and scarring, cholesteatoma, damage to the ossicles, mastoiditis, and labyrinthitis (Bluestone et al., 1990; Chapin and Bluestone, 2009). Goudy et al. (2006) found that 6 of 101 patients with clefts who were 8 years to 25 years old at the last follow-up had cholesteatomas. Twenty-five percent of the 101 patients had long-term conductive hearing loss. It is not surprising that children with congenital VPI in the absence of an overt cleft also show a substantial incidence of otopathology and hearing loss and require the same careful monitoring as children with clefts (Sheahan et al., 2004).

On the positive side, several reports have noted the eventual disappearance of middle ear disease and conductive hearing loss as youngsters with clefts grow older (Goudy et al., 2006; Gould, 1990; Skuladottir et al., 2015). These findings underscore the importance of following young patients over a long period of time. In

one study of children without clefts who had otitis media and hearing loss who had shown a relationship between level of hearing loss and deficits in language development at 27 months of age, that relationship was no longer evident by the time they reached school age (Zumach et al., 2010).

Although the type of hearing loss reported in children with clefts has typically been a mild to moderate intermittent conductive loss, several studies reported high-frequency losses (e.g., 8,000 to 20,000 Hz range) (Ahonen and McDermott, 1984; Handzic-Cuk et al., 1996; Manning et al., 1994). McDermott et al. (1986) concluded that persistent middle ear disease alone is sufficient cause for loss of auditory sensitivity in the high frequencies. Other investigators have suggested (1) that high-frequency sensory hearing loss in patients with OM could be the result of toxic metabolites passing across the round window membrane into the inner ear (Goycoolea et al., 1980; Paparella et al., 1972, 1984) and (2) that conductive high frequency losses could result from anatomical changes in the inner spaces (Strohm, 1986; Tonndorf and Pastaci, 1986). Tierney et al. (2015) interviewed the parents of 37 children with clefts regarding their child's middle ear problems, together with 22 of the children themselves. The interviews uncovered frustration, anger, anxiety, and other emotions linked to the ear disease and its consequences. These investigators advised that the early counseling given to the parents of children with clefts include specifics about the likelihood of ear problems and, of course, the need for routine monitoring and hearing tests.

For obvious reasons, there has been long-term interest in the questions of whether ear disease and hearing loss decrease after closure of the palate, and, secondarily, whether the technique of closure makes a difference in subsequent status of the ears. The first question is difficult to answer because the effects of palatoplasty on ear health cannot be measured independently of the effects of growth and time. Evidence indicates that ear health is more substantially improved when the palatoplasty includes reconstruction of the levator palatini in comparison to palatoplasty without such reconstruction (Hassan and Askar, 2007).

### Current Trends in Treatment

Most of the more recent literature has continued to point to the higher occurrence rate of otitis media in children with clefts as compared with children without clefts and supported the need for myringotomies and ventilating tubes (Kobayashi et al., 2012; Kwan et al., 2011; Merrick et al., 2007). However, this treatment approach is not without controversy. Otolaryngologists have been questioning whether myringotomies and ventilating tubes are always necessary since at least 1992 (Robson et al.). Tuncbilek et al. (2003) reported normal hearing in 63% of individuals with clefts whose treatment regimen had *not* included myringotomy and ventilating tubes. These investigators thus questioned whether myringotomy and ventilating tubes should really be routine for children with clefts. Ponduri et al. (2009) conducted a retrospective literature review (although only on 42 articles) on placement of ventilating tubes in children with clefts and concluded that there was "insufficient evidence on which to base the clinical practice of early routine grommet placement." Their concern was the poor quality of the studies and the potential damage to the tympanic membrane. Interestingly, the article by Ponduri et al. (2009) was published at the same time

as a large study of Chinese patients with unrepaired palates ranging in age from younger than 1 year to adulthood (Zheng et al., 2009). At least 30% of the patients in each age group had hearing loss and abnormal tympanometric results. Zheng et al. urged palate repair and tube insertion at an early age to avoid interference with language and educational development.

Concern about the effects of hearing loss on language and educational development in children with clefts is not new, but several studies since the 1990s have provided alerts for related concerns. Cheour et al. (1999) and Ceponiene et al. (1999, 2000) reported evidence of dysfunction of the auditory cortex in youngsters with clefts, a problem they did not see as directly attributable to middle ear disease. In 2008, Maenpaa et al. reported problems in temporal processing acuity (transmodal processing of audiotactile, visuotactile, and audiovisual information) in children with clefts and discussed the potential role these problems could play in the development of learning disabilities. Yang et al. (2012) reported structural abnormalities of the central auditory pathway in infants with clefts, specifically smaller volumes of the left thalamus and left auditory cortex. All these studies fit into the overall picture that has developed regarding learning disabilities and structural anomalies of the brain in children with clefts.

### Summary for Speech-Language Pathologists

Routine use of myringotomy and tubes in infants with clefts is not quite the automatic treatment decision today that it first became in the 1960s, but many treatment centers and individual otolaryngologists do insert tubes in the first few months of life (sometimes at the time of lip repair to reduce the number of anesthetic experiences the child undergoes) if there is evidence of middle ear fluid. Other clinicians prefer a more conservative approach with a period of observation and perhaps medication before resorting to tubes. What matters most is that all caregivers recognize the high likelihood of otitis media and hearing loss and share that information with the parents, who are after all the first line of defense. SLPs should be particularly interested in a large study (N = 370) of hearing and speech and language function in children with clefts in Germany (Schonweiler et al., 1999). These investigators reported that phonology, morphology, syntax, vocabulary, and language comprehension were predominantly affected by hearing status, not by the type or extent of cleft or VPI.

## SOCIAL, PSYCHOLOGICAL, AND EDUCATIONAL ISSUES

Common sense tells us that it must be difficult to have a facial difference that marks you as someone who is not quite like everybody else. Youngsters want to "blend in," which is difficult if you have a facial difference. The social and psychological problems that clefts can cause affect the parents and grandparents as well as the patients themselves, and the problems are not easily eradicated. All professionals involved in the care of children with clefts have, or should have, an overriding focus on "quality of life." They know that the most important goal is to get each youngster up to the quality of life he or she would have had without a cleft. That is why, on interdisciplinary teams, it is not just the psychologist or social worker who focuses on the psychological well-being of the patient and family. The entire team is focused on producing a happy young adult.

The amount of published research on psychological, social, adjustment, and educational issues for children with clefts and their families could be viewed, historically, as a mushroom cloud. In mid-20th century, there were a handful of psychologists studying these topics. In the 1970s and 1980s, the number of workers and the number of published studies rapidly expanded, and these numbers have grown more exponentially than numerically in the late 20th and early 21st centuries. Extensive lists of references are found in Kapp-Simon and Gaither (2009) and Peterson-Falzone et al. (2010).

Many of the older studies on the psychological and behavioral effects of clefts pooled subjects with cleft lip with or without cleft palate and cleft palate only. This pooling is particularly bothersome because patients with cleft palate only are far more likely to have other congenital anomalies than are patients with cleft lip with or without cleft palate and are also more likely to have a syndromic diagnosis (see Peterson-Falzone et al., 2010). Some of the more current studies have continued to pool data across cleft types and even included patients with other types of congenital craniofacial defects (e.g., craniosynostosis) in the effort to gain larger numbers of patients in the nonspecific category of "facial differences." Notwithstanding these and other methodological problems, the body of literature cited by Kapp-Simon and Gaither (2009) and Peterson-Falzone et al. (2010) has shown the following:

1. Individuals born with clefts generally test within the normal range on various forms of intelligence tests.
2. There is no one-to-one relationship between the severity of the cleft and the level of distress manifested by the parents.
3. Similarly, there is no one-to-one relationship between the amount of facial disfigurement and the child's self-esteem or adequacy of social development.
4. Children with clefts do have a higher than average tendency to have reading disabilities and learning problems, and their school achievement level is often one full grade below chronological age (Broder et al., 1998; Richman, 1980; Richman et al., 1988).
5. Imaging studies have found abnormalities in brain structure in individuals with nonsyndromic clefts (Nopoulos et al., 2000, 2001, 2002, 2007; Richman and Nopoulos, 2009; Rosen et al., 2011; van der Plas et al., 2010); researchers are beginning to relate these changes to function, such as behavior (Nopoulos et al., 2005, 2010) and speech (Conrad et al., 2010).

## Issues by Stages in Life

How well children with clefts do in life is determined by many factors, including how well their parents do when those children are born. To understand all the influences on the social, psychological, and educational well-being of patients, it is best to take a chronological approach. Table 6-2 summarizes points of concern along the time line from prenatal to adult life.

### Parents

Getting any child successfully launched in life is a huge responsibility that requires diligence, energy, family support, resilience, a sense of humor, and long-term

**TABLE 6-2**

*Psychosocial Concerns According to Developmental Stages of Patients*

| DEVELOPMENTAL STAGE OR AGE BRACKET | AREA(S) OF CONCERN | SPECIFIC TOPICS |
|---|---|---|
| Prenatal | Adjustment to the impending birth of a child with a defect | — |
| Birth, perinatal period | Adjustment to the birth of a child with a defect | Parent-infant bonding, feeding issues, reactions of other family members, dealing with reactions of others in the community; stress and financial demands of surgery* |
| Toddler years | Parent and family adjustment to multiple demands | Parents' attempts to protect the child from adverse reaction from extended family and community; child's normal attempt to assert self before the realization of personal difference; stress of medical interventions |
| Preschool years (ages 3-6 y) | Self-concept; peer relationships; cognitive development | Unavoidable realization of facial difference for child; judgments by peers and teachers; interruptions from medical interventions; possible learning disabilities |
| Early school years (ages 6-10 y) | Self-concept; peer relationships, school adjustment, achievement | — |
| Preteen years (ages 10-13 y) | Self-concept, peer relationships, school achievement | Judgments by peers and teachers; interruptions caused by medical interventions; possible learning disabilities |
| Teens | Self-concept, peer relationships, school adjustment, achievement | Final phases of physical management (e.g., orthodontics, surgery) |
| Adults | Social interactions, life partners, employment | — |

From Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP: *Cleft palate speech* (4th ed). St. Louis: Mosby, 2010.
*Including indirect costs of factors such as absences from work, travel, child care for siblings, and housing.

planning. All parents-to-be wait in anxious anticipation for the new baby who will be, undoubtedly, both a wonder and a challenge. When that baby has a cleft palate or other birth defect (particularly if undetected prenatally), the parents feel frightened and cheated. What went wrong? Was it somehow their fault? The parents also sense their new job will demand more of them than they had expected. They quickly realize that they will need to find sources of care. In these days of online "everything," they will spend a lot of time in front of a computer screen, but they also need to know that they can pick up a telephone and make

a single call (1-800-24CLEFT) that will lead them to all kinds of help. This is the number for the hotline of the Cleft Palate Foundation, the public branch of the American Cleft Palate-Craniofacial Association (ACPA). They will be referred to a nearby team that has met the ACPA standards, and information on the brochures published by the Foundation will be sent to them.

> The Internet is not peer reviewed. Anyone can say anything. It is largely composed of opinion, not science or fact. This is a great danger to families desperate for information.

Most cleft palate or craniofacial teams have a nurse, pediatrician, or speech pathologist who can help solve feeding problems. Most teams also have identified parents who are willing to meet with the new parents to give support and ease their anxiety. Much of the early stress for parents stems from concerns over (1) how this child is going to feed and thrive, (2) how they are going to present their newborn to others both inside the family (particularly grandparents) and outside the family (friends, neighbors), (3) what the future problems may be (speech, teeth, social acceptance), and (4) how they are going to meet the needs for frequent medical visits without giving short shrift to the rest of their lives, particularly if they have other children. Fulfilling all these needs can be exhausting, and parents need to know that others have been through the same challenges. That is why meeting with other parents can be very helpful.

Various forms of online social media have become a major source of information (although not scientifically vetted information) and support for parents. They can connect with other parents, find help on feeding and social-reaction issues, vent their frustrations and fears, and generally gain the comfort of knowing there are others "out there" who know what they are going through. Further, several social media sites provide the means for setting up private groups so that parents are not posting their cleft-care frustrations to just anyone who uses that particular site.

> Unfortunately, there are still reports of infants with nonsyndromic clefts being gavage-fed or tube-fed in the birthing hospital and sent home with antiquated information on feeding (e.g., relying on tube feedings).

In a multistate, multicenter examination of mental health status and aggravation level among mothers of nonsyndromic children with clefts, Dabit et al. (2014) identified some unsurprising factors associated with increased poor maternal mental health: less education, lower household income levels, multiple children, and a tendency to rate both their own health and that of the children lower than the mothers of children exhibiting either moderate or low levels of aggravation. The major point of the report was to urge that "brief screeners" for mental health and parenting be administered during routine clinic visits to identify at-risk caregivers.

### Prenatal Diagnosis: Anticipation of a Child With a Cleft

How parents and extended families deal with the birth of a child with a cleft or other congenital craniofacial anomaly is certainly influenced by prenatal diagnosis

and counseling. We would like to think that early provision of information (before the baby is born) allows families to deal with all aspects of the situation with more equanimity: early care and feeding, family reactions, sibling response, and so forth. Prenatal diagnosis of a cleft lip with or without cleft palate by ultrasound imaging is now common. Prenatal detection of cleft palate only is still a work in progress, with a few cases having been picked up because the cleft was part of a multimalformation syndrome or sequence (e.g., Pierre Robin sequence).

Geneticists, psychologists, and social workers have accumulated a body of knowledge on (1) how best to give this information to the parents-to-be; (2) what they, as team members, should be ready for in terms of family reactions; and (3) seeking later feedback from parents regarding how helpful their counseling was or was not, and how they should change their approach. Of course, the information conveyed to families must be presented in "user-friendly" terms and delivered in consumable doses (e.g., not everything at one time). Information often has to be repeated and should be available in printed form so the family can study it at home. Parents report needing to be prepared for feeding and early health care issues (Young et al., 2001). If they do not actively seek to meet other parents, the counselor will suggest this. The psychologists and social workers (or whoever is doing the prenatal counseling) will talk with the parents about preparing their friends and extended family for the birth and suggest ways of handling questions. Of supreme importance will be the offer of consistent, easily available help and support once the baby is born.

### Infants and Their Parents: The Initial Reaction

As silly and inappropriate as it seems now, there was a time when birthing hospitals delayed showing a child with a cleft to the parents. Today, fathers are not kept out of delivery rooms, and babies go straight from the womb into the arms of the mother. Delays in seeing the baby heighten anxiety, whereas being able to hold the infant and keep him or her nearby allows parents to realize that the baby is more like a normal child than not and that they can do normal things such as bathing, cuddling, and comforting. The parents in the study by Young et al. (2001) reported wanting to be shown all the things about their baby that were *normal*. Parental reactions to first learning of the baby's cleft can include disbelief, shock, anger, guilt, depression, resentment, grief, rage, frustration, fear, anxiety, protectiveness, and stigmatization. They need time, moral support from family and friends as well as from the professionals caring for the child, and accurate information in order to adjust. It may help when they learn that a cleft lip or cleft palate is "fixable," although not totally "erasable." Some parents fear that their child may be "retarded."

> References for published studies relevant to early parental reactions may be found in Peterson-Falzone et al. (2010) and also on the website associated with this book.

The team (or the child's pediatrician) will need a complete examination of the child by a geneticist/dysmorphologist as soon as possible. Although all the professionals seeing the child and family will be anxious to reassure the parents about the child's overall health and chances in life, it is best to do so *after* the child has

been seen by the geneticist/dysmorphologist. At some point, this same professional will take a family history.

Evaluation of the baby and family members by a geneticist/dysmorphologist in the early weeks of life is important for accurate and complete diagnosis, treatment planning, and counseling for the family. In addition, keep in mind that some congenital conditions do not become apparent until later in life, so access to evaluations by a geneticist/dysmorphologist should always be available to children and families.

When there is a cleft in one of the parents, the parents are significantly more likely to feel a sense of guilt than are parents without clefts, but they report significantly less anxiety in comparison to parents without clefts (O'Hanlon et al., 2012).

With the help of the team and other parents, the new parents should be less likely to withdraw from social contacts and less likely to conceal the baby from public view. Interestingly, Sank et al. (2003) found that mothers of infants with visible clefts reported higher perceived social support than did mothers of children with nonvisible clefts. In other words, the visibility of the cleft seemed to elicit more concern and support from family and friends. Of course, parents of infants with cleft palate only could conceivably delay in letting their child's condition become known to friends, thus depriving themselves of "someone to talk to" when they most need it.

In 1979, MacDonald, a parent of a child with a cleft, wrote, "Parents take their cues from professionals." Seeing the professionals smile at the baby, cuddle the baby, and so forth can ease parental anxiety. SLPs quite naturally take advantage of this "modeling" strategy, by engaging in interactive cooing (and babbling, when it is time) and making funny faces to the baby to see whether he or she responds. The positive, enjoy-your-baby model provided by caregivers is as necessary as the provision of contacts with support groups, provision of accurate information, and always having the time to listen to questions and concerns. In reports from parents' interviews or questionnaires, two consistent desires expressed by parents are the need for more compassion from professionals and more time to discuss their concerns (Strauss et al., 1995; Young et al., 2001). Nelson and Kirk (2013) interviewed parents of children with clefts across a wide age range (20 weeks to 21 years) and found that one of the key reasons given for dissatisfaction with treatment was a lack of moral support from professionals.

### Dealing With Early Demands

Just at the time that parents and infants are bonding to each other, the parents of infants with clefts are dealing with multiple early needs. In addition to establishing a successful feeding routine, there is often the stress of necessary early diagnostic evaluations (pediatric, audiological, otological, genetic, and others) and the stress associated with early stages of treatment. Many babies in the United States undergo lip surgery at approximately 3 months of age and palate surgery at approximately the age of 12 months (some earlier, some later). If the baby is being seen by a team that uses infant orthopedics, there will be frequent clinic visits in the first few weeks and months of life.

> For the family, the operations and the perioperative periods can be a time of conflict and certainly of extra demands on finances and support. In the early days and weeks of the child's life, although the parents know that the baby is small and vulnerable, they often inquire about the possibility of immediate surgery to correct the visible defect. When it is actually time for the lip surgery, parents typically are pleased that the defect is finally going to be "fixed," but because they will have had the child at home for 2 or 3 months before surgery, they may find that they are actually reluctant to see the appearance of their baby's face altered. It is not unusual for parents to admit a little sheepishly that they "don't really want to see his smile change."

Hospitalization of the baby is now a more "family-friendly" experience, with a parent sleeping near the baby and serving as the primary caregiver and the hospitalization time kept to a minimum. Nevertheless, the hospitalization and surgery can amplify the parents' feelings of guilt (e.g., "We did this to him. If it wasn't for us he wouldn't have to go through this."). Hospitalization for the palate surgery takes a little longer than for the lip surgery, and the feeding method may be temporarily altered (also upsetting to the baby). In addition, the baby is older, more aware of his or her surroundings, and more tightly bonded to the parents. The parents must be carefully informed of what to expect, and moral support must be consistently available to them.

Among the points of information that should be covered with the parents preoperatively is the use of arm restraints after surgery to prevent the baby from sticking his or her fingers or other objects into the mouth. Arm restraints used to be employed religiously after both lip and palate surgery, but they have become less popular over time.

Fortunately, there is evidence that the stresses of the early demands do not last forever. In a study of 40 babies with clefts and 45 controls, Habersaat et al. (2013) found that babies with clefts at 2 months of age were more difficult and less cooperative with their mothers than were babies without clefts. The investigators attributed this difference to the stress engendered by treatment. However, at 12 months of age, no differences were found between the infants with clefts and those without clefts in attachment security. The children with clefts and their mothers were doing as well as families without clefts in terms of the mothers' mental health, mother-child relationships, and quality of attachment. These investigators believed that a potential reason for this outcome was the pluridisciplinary support that the families of infants with clefts had received. Many years earlier, Speltz et al. (1994) reported similar findings: negative parent-infant interactions that were apparent in the early weeks of life, specifically at feeding times, had resolved by 12 months of age. Maris et al. (2000) also found a pattern of resolution over time. That group examined mother-infant attachment in three groups of babies (cleft palate only, cleft lip and palate, controls) at 12 and 24 months of age. Although the babies with cleft palate only showed a lower level of attachment at 12 months, by the age of 24 months, most of the babies with clefts were showing secure maternal attachments.

### Toddlers

Although toddlers are not "short adults," they assert themselves in new ways every day. They are walking and talking and making their needs and feelings known.

Researchers in cleft care have been evaluating toddler motor development, cognition, comprehension, and expressive language abilities for many decades. Many of the studies published in the 1970s, 1980s, 1990s, and the early part of the 2000s demonstrated delays, particularly in children with cleft palate only (see Peterson-Falzone et al., 2010, Chapter 13, for historical references).

After conducting a large study (both cross-sectional and partially longitudinal) on 180 infants and toddlers with clefts up to 24 months of age, Kapp-Simon and Krueckeberg (2000) remarked that the number of children at risk for developmental problems during the second year of life was "larger than expected." These investigators urged more active intervention in the early childhood years, thus echoing the plea of other investigators in earlier studies. In retrospect, it is tempting to wonder whether some of the delays found in these studies were related to the differences in brain structure that began to be recognized in the early 2000s (Nopoulos et al., 2000, 2001, 2002).

Depending on the treatment regimen of the team caring for the child, another surgical procedure may be performed during the second or third year of life. Some children may have late palatal surgery because of failure of the first repair, a delay in repair as a result of medical problems, or other reasons. When "late" palate surgery is performed, the parents fear that the normal falls a toddler experiences while learning to walk and run could damage the repair, and may feel it necessary to curb their child from his or her normal activity level. In addition to palatal surgeries, other procedures (e.g., myringotomies, even though these do not require hospitalization or isolation from parents) interfere with the toddler's life and require going to scary places (hospitals, doctor's offices). These interruptions could conceivably be related to lags in development.

Parental stress was shown to be related to adjustment and behavioral problems in toddlerhood and in the preschool years in several studies of children with craniofacial anomalies compared with control groups (Pope et al., 2005; Speltz et al., 1990; Speltz et al., 1993). Only a portion of the sample in these studies was composed of children with clefts; in the case of the 2005 study by Pope et al., 30 of the total number of 47 patients had clefts. However, the findings alert caregivers to the need to assess parenting stress in all families and try to offer assistance.

### Preschool Children

The new challenges in the age range of 4 to 6 years will likely include the child's perception of his or her own facial differences, if he or she has not reached this step already. There are also the challenges of an expanded social world. The first experience with a school situation may be the child's first experience with querying looks, questions, and teasing from other children. A further surgical experience may occur at approximately age 6 years, specifically a lip and nose revision. (The timing of this procedure is determined by the desire not to interfere with the major growth centers of the nose.) Both the parents and the child must be prepared for the fact that this surgery will *not* erase all signs of the original cleft.

In a tutorial for plastic surgeons, Kapp-Simon (1995b) stated that some toddlers and preschoolers should not have elective surgery (such as a lip-nose

revision) until the child can be a more active participant in the decision making. She urged that the emotional characteristics of the child and the strengths in the parent-child relationship be evaluated before such surgery is scheduled. In a study of 20 children ages 4 through 7 years with a variety of types of congenital facial anomalies, Fisk et al. (1985) found "increasing emotional indicators" in those children who had had more or later operations. It is no surprise that Tyler et al. (2013) found evidence of separation anxiety in 24% of children with clefts between the ages of 4 and 9 years, compared with U.S. child population estimates of 3% to 5%.

Members of the craniofacial team should help the child and family prepare for preschool by suggesting answers that he or she can use in responding to questions and unkind remarks from children. Once querying children are given a straight answer to a question, they are typically "disarmed" and ready to move on to another topic. The more success a child with a cleft has in handling early confrontations, the less likely he or she will be to exhibit social withdrawal or reticence. It is also a good idea to remind the child that everybody gets teased about something.

Historically, the earliest age at which researchers attempted to measure intelligence in children with clefts was in the preschool years. However, as pointed out by Richman and Eliason (1982), intellectual assessments during this period should be interpreted with caution, especially when highly verbal measures are used. A chain of pitfalls is possible in this regard: A child with a delay in phonetic development resulting from early structural constraints (e.g., the unrepaired cleft) can exhibit a secondary delay in phonological and language development. Given the fact that preschoolers with clefts are coping with new challenges in a new environment, it is not surprising that behavioral problems begin to become apparent.

Multiple studies of social skills and behavior have been conducted in youngsters of all ages with clefts (see Peterson-Falzone et al., 2010, chapter 13 for historical references). In the toddler and preschool years, these studies have consisted primarily of parent and teacher questionnaires and behavioral rating scales, although Krueckeberg et al. (1993) included a social knowledge interview of the child himself or herself in their study of social skills in 3- to 6-year old children with a variety of craniofacial anomalies (CFA), including clefts. In this study, boys with craniofacial anomalies fell in the average range for self-perception, but girls rated themselves above average. The investigators conjectured that these children may not have yet encountered the stigmatizing reactions to facial appearance that can lead to lowered to self-esteem and negative social interactions. This study became partially longitudinal when the investigators (Krueckeberg and Kapp-Simon, 1997) reevaluated a small subgroup of their 1993 subjects and found that 31% were exhibiting global behavior problem scores in the clinical range (meaning clinically significant) as rated by either teacher or parent report. Krueckeberg and Kapp-Simon (1993) examined the parenting stress and parenting style and the social networks of parents with 30 of the families involved in the 1993 study by Krueckeberg et al. These investigators found no differences between the parents of children with craniofacial anomalies and control groups in parenting stress, parenting style, or social network characteristics. However,

parents of children with visible defects found their social support networks more helpful. In the group with craniofacial anomalies, parenting stress predicted children's level of social skill. This finding is not surprising, but it reminds us of the importance of staying tuned to the level of parental stress.

### School-Age Children

Self-concept, self-confidence, social acceptance, school achievement, and conflicts with parents are issues in the life of virtually every school-age child. It is not difficult to conceptualize how the presence of a cleft could complicate any one of these areas. What is most worrisome is (1) children with clefts have a higher than average likelihood of reading disabilities and learning problems; (2) their school achievement level is often one full grade below chronological age (47% of children with clefts function below grade level, and 36% demonstrate reading disabilities) (Broder et al, 1998; Richman, 1980; Richman et al., 1988); and (3) even nonsyndromic individuals with clefts may have anomalous brain structure (Conrad et al., 2010; Nopoulos et al., 2000, 2001, 2002, 2007; Richman and Nopoulos, 2009; Rosen et al., 2011; van der Plas et al., 2010).

As stated by Richman et al. (2012, p 387): "For many children with cleft, delayed speech and language development has been shown to be related to subtle auditory memory deficits, expressive language difficulty, and sometimes more severe central language impairment, which can result in long-term reading deficits (Richman 1980) and other learning disabilities (Richman et al., 2005)."

No teacher or counselor would want to emphasize any of these findings to a youngster with a cleft or to the child's family. Rather, the emphasis should be on *prevention* of learning and reading problems by intervention. Careful assessment of visuomotor skills, visual memory, verbal labeling, comprehension, phonological development, and expressive language skills should be carried out in the preschool years and repeated at intervals throughout the school years. Both speech-language therapy and intervention for learning and reading problems should be instituted, when indicated, and vigorously maintained to raise the youngster up to his highest level of potential.

In a very early study, Richman (1976) looked at both behavior and achievement in school-age children (9 to 14 years old) with clefts compared with a matched control group. School achievement was lower for both boys and girls with clefts (even though intelligence quotients were matched between groups) and lower for the boys than for the girls. Classroom teachers complete a behavior problem checklist for each child. The children with clefts were rated as showing excessive inhibition of impulses. Although inhibition of impulse is something we expect children to develop gradually as they mature, children with clefts seem to show greater inhibition (sometimes equated with shyness, social withdrawal, and so forth) than considered normal for chronological age. Clinicians and researchers have attributed the increase in inhibition to an unconscious attempt by the child to avoid calling attention to himself or herself or to avoid situations that give rise to negative responses from others. Richman (1976) pointed out that although impulse inhibition may be a way of coping with negative social responses from peers, in the competitive classroom environment the inhibited child may meet with less success than his or her peers.

The link between problems in social adjustment and problems in school achievement has reappeared in multiple studies on children with clefts since this study was published. The behavioral characteristics of passivity and social withdrawal or social reticence were frequently reported in children with clefts in studies throughout the 1970s, 1980s, and 1990s. However, more recent studies paint a somewhat different picture. See the studies by Brand et al. (2009), Collett et al. (2012), Feragen et al. (2009), and Klein et al., (2014) that are discussed later. SLPs should be especially interested in a 2011 study by Chapman in which she compared the reading skills, speech production abilities, and language abilities of 28 5-year olds with clefts with those of matched control children. She found a statistically significant correlation between early reading skills and speech production abilities, as well as between early reading skills and language abilities. The children with the most severe speech problems were the children with the poorest performance on the reading test. Chapman emphasized the need for early intervention. In a similar vein, Lee et al. (2015) assessed 15 nonsyndromic youngsters with cleft lip and/or palate in the age range of 6 to 8 years and once again reported problems in phonological awareness and spelling skills, the kinds of problems we know can lead to long-term problems in reading and learning.

Although having a cleft can complicate some aspects of school adjustment, achievement, and social behavior, a cleft does not have to mean inevitable failure, frustration, or unhappiness for the child and parents. Two publications by Collett et al. (2010, 2012) reported data from a longitudinal study of a group of children with clefts and a large group of controls. These children were followed from infancy through the age of 7 years. Contrary to what had been published in many other studies on language problems in children with clefts, Collett et al. (2010) found no significant group differences in language at ages 5 and 7 years, and at 7 years the children with clefts scored significantly higher than controls on measures of early reading skills. However, 56.5% of the children with cleft lip and palate and 40.9% of those with cleft palate only were in special education placement, as compared with 19.3% of the controls; the most frequently reported special education classification for all groups was communication disability, followed by specific learning disability.

Collett et al. (2010) pointed out that demographics constituted one of the predictive dimensions in this study: most of the children were from white, non-Hispanic, middle-class families. Other predictive variables were early mother-child interactions during teaching and feeding tasks and cognitive development scores at age 24 months.

In the later study, Collett et al. (2012) looked at psychosocial functioning in an expanded number of both children with clefts (N = 93) and controls (N = 124). Their data came from questionnaires mailed to families that included checklists for child behavior and social competence, in addition to an inventory of parental stress. These investigators found minimal differences in psychosocial function between the youngsters with clefts and controls and minimal differences between the parents of the children in the two groups. This was a population-based study, as opposed to a study using patients from a clinical database. These investigators remarked: "Psychosocial differences may be less apparent in

population-based versus clinic-based samples. Alternatively, social-emotional deficits may become apparent in older school-aged children, making the preschool and early elementary school years an optimal time for preventive intervention" (p 397).

School-age children have a lot to say about themselves and thus constitute a source of valuable information for researchers. Feragen et al. (2009) obtained 10-year follow-up questionnaires completed by 722 10-year-old children with clefts and found that psychosocial resilience was associated with adequate emotional functioning, high satisfaction with appearance, and a lower frequency of reported teasing. In a later study, Feragen and Stock (2014) included child interviews, not just rating scales of behavior, as they derived data on psychosocial adjustment in children with multiple anomalies. Brand et al. (2009) conducted a cross-sectional study of 32 youngsters with clefts who were between the ages of 6 and 16, comparing the findings in those youngsters with those in 34 controls. These investigators looked at emotional problems and conduct problems or hyperactivity and found that the youngsters with clefts did not differ from the control group with respect to any of these parameters, yet they were six times more likely to self-report problems with interactional competencies. These findings remind us of the importance of listening to the child, rather than just observing or rating the child's behavior. Another encouraging set of results for school-age youngsters came from Klein et al. (2014) in which the mothers of children with clefts in the age range of 9 to 14 years were found to be appropriately concerned about their children's peer relationships and thoughtful in devising strategies to manage social challenges successfully.

### Adolescents

It is unlikely that anyone who has ever parented a teenager would describe that task as problem-free. The need for autonomy that appears in toddler years appears again in the early teens and causes just as many parent-child battles. Parents strive to "support without smothering." Kapp-Simon (1995a) pointed out that the normal developmental tasks of adolescents include individuation from family, development of a sense of personal identity, and establishment of satisfactory peer relationships.

Not surprisingly, many studies have demonstrated the importance of satisfaction with appearance for teens and the role this satisfaction plays in self-concept and social relationships. Kapp-Simon (1995a) made specific suggestions for helping teens handle teasing, drawn from her social skills training program (Kapp-Simon and Simon, 1991). In this program hypothetical situations are presented to the youngster on videotape for rehearsing responses that are direct, honest, and unlikely to stimulate further teasing.

Teenagers are old enough to participate in decisions about any further treatment procedures (Kapp-Simon, 1995a; Strauss, 2001). When it comes to proposed surgical or orthodontic procedures, he or she may volunteer, "I'm satisfied with the way I look," only to have the parent stress the need to follow "the doctor's" recommendation. Both caregivers and parents must be careful that the youngster has been given accurate, complete information about what "the problem is" and what is being proposed for treatment. In addition, some teenagers object to more

speech therapy. Unless the teen himself or herself agrees that it is necessary and believes that it can make a difference in his or her life, therapy is destined to be useless.

Strauss et al. pointed out in 1988 that teenagers with craniofacial anomalies are not necessarily out of the woods by virtue of either maturation or treatment (orthodontics, surgery) by the time they are on the brink of adult life.

We would like to think that things have changed for the better in the past 3 or 4 decades. In a large study of 145 families of adolescents with clefts, Berger and Dalton (2009) found little evidence of psychosocial adjustment difficulties in either the teens or their mothers. These investigators felt that their results highlighted the importance of timely interventions and seeking multiple perspectives in the clinical management of a child with a cleft. These same investigators (Berger and Dalton, 2011) later went back to look at factors predicting psychosocial adjustment in both the teenagers and their mothers. In the adolescents, psychosocial adjustment was predicted by their social experiences and the well-being of their mothers. For the mothers, adjustment was predicted by their own acceptance of the situation (rather than denying problems) and by the coping strategies they used. In this report, greater adjustment difficulties were found in boys than in girls. Boys tended to use the avoidant coping strategy of "blaming others." These findings were in opposition to those of Kapp-Simon and Dawson (1998), who found that adolescent girls exhibited more elevated levels of behavior problems compared with boys.

Overall, there are conflicting reports on whether boys or girls exhibit the most problems in psychosocial adjustment. Adjustment to facial differences may be more difficult for girls, but boys tend to "externalize" more by blaming others and acting out. There is ample evidence that both school-age children and teenagers exhibit dissatisfaction with physical appearance (Slifer et al., 2004; Thomas et al., 1997) and associated teasing from peers (Hunt et al., 2006). In a 2007 follow-up study of 129 young people with clefts who were between the ages of 8 and 18 years, Hunt et al. found the most significant predictor in parental reports of psychosocial impairment in their children was a history of teasing related to facial appearance. Kapp-Simon et al. (2005) cautioned that use of social withdrawal as a protective coping mechanism in adolescents may increase the risk of teasing and have an impact on overall levels of adjustment.

The 2013 report by Tiemens et al. on teenage girls with clefts was disheartening for its documentation of continued problems with teasing, bullying, and stares, but it was interesting in that the subjects (N = 7) worked at ways to reduce the impact of social stigma through coping strategies and social supports and by seeking further reconstructive surgery. Although the small number of subjects in this study limits the impact of the conclusions, this report offers a hopeful note that teens are learning to handle their challenges.

Teenagers' satisfaction with speech also plays a role in overall satisfaction with treatment (Havstam et al., 2008; Hunt et al., 2007; Richman and Millard, 1997). Havstam et al. (2011) interviewed adults with clefts in their 20s and 30s to learn what strategies they used as teenagers to cope with the social results of less than normal speech. Many of their responses would qualify as forms of withdrawal. It

is no surprise that, in the 2012 study by Dzioba et al., adolescents with clefts who exhibited VPI in speech also exhibited more communication apprehension than did youngsters without VPI. It is safe to assume that dissatisfaction with their own speech contributes to a tendency toward social withdrawal in many youngsters.

In terms of coping strategies, patient groups who self-report the use of "approach-oriented" rather than "avoidance-oriented" strategies in social contacts seem to do better in psychosocial adjustment (Baker et al., 2009). The same finding is true both in parents of children with clefts and in teenagers with clefts. In the study by Baker et al. (2009), negative outcomes both in parents of children with clefts and in teenagers with clefts were not high, and the participants reported high levels of social support. The outcome equation seems to be "approach strategy" + high levels of social support = lower psychological stress and better adjustment.

Two other reports demonstrate that more work remains to be done in launching teenagers with clefts successfully in life. Snyder and Pope (2011) conducted a retrospective chart review on a large group of children 2 years to 18 years old who had a variety of craniofacial anomalies, including 144 individuals with cleft lip, cleft palate, or both. The behavioral data came from a behavioral checklist filled out by the parents, guardians, or a close family member. The group with cleft lip and palate showed elevations in social problems and deficits in social, academic, and activity competencies. A later report of 511 adolescents with clefts in Sweden (Persson et al., 2012) found that these teens had significant deficits in their educational achievements in comparison with unaffected peers. Their grade-point averages were lower, and they had higher odds of not graduating.

> The results were reminiscent of a very old study by Demb and Ruess (1967) on high-school dropout rates in teenagers with clefts in the Chicago metropolitan area.

It is a little more encouraging that Hall et al. (2013), in interviews and autobiographies elicited from 17 youngsters with clefts who were 8 to 17 years old, found that those youngsters tended to reflect on how they "just got on" with their lives and felt that their treatment procedures did not particularly affect them. These observations reflect resilience, something that psychologists want to see in young people with clefts.

## Adults

It is unlikely that many SLPs functioning outside a craniofacial team will encounter adults with clefts. This section is offered only in the interest of providing a perspective. Strauss (2005) stressed the need for teenagers and adults with clefts to have social support systems and social interactions. Based on the literature and from what he himself learned in interviewing patients, he also stressed the need for good outlets for talking to others and for "unconditional conversations" with people who accept the individual for exactly who he or she is.

Mani et al. (2010) studied 86 Swedish adults with repaired unilateral cleft lip and palate and found lower than normal values on a self-report mental health scale. Men were affected more negatively by their clefts than were women in terms of emotional function in daily life. Younger adults (ages 20 to 32 years)

were more negatively affected than older adults (ages 33 to 47 years) on several of the sub-subscales (social function, physical aspects, emotional aspects, mental health, vitality, bodily pain, general health) of a questionnaire assessing health-related quality of life.

In a study of French adults with repaired clefts, Danino et al. (2005) found a significant delay in educational achievement and in living on their own. Income levels were substantially lower in comparison to a control group. These investigators concluded that the patients with clefts, as a group, had a significant delay in the process of becoming independent.

It should be noted that, in the 1997 study by Thomas et al., satisfaction in facial appearance was lower in teenagers (age 10 to 15 years) than in individuals older than 20 years, possibly reflecting an effect of maturation. However, Cheung et al. (2007) found exactly the opposite: individuals with clefts who were between 10 and 16 years of age showed higher self-esteem than did individuals 17 to 40 years old. It is tempting to wonder whether the younger group had better moral support and interventional counseling or more advanced treatment (or both) than the older group.

The subject groups in the neuroimaging studies of Nopoulos et al. (Nopoulos et al., 2000, 2001, 2002, 2005, 2007; Richman and Nopoulos, 2009) consisted primarily of adult men. However, it is important to note that we do not yet know the true significance of their findings in terms of emotional health or functional abilities. It is difficult to obtain data on adults with clefts because once treatment is completed these patients move on with their lives, often literally, so they are no longer accessible to the team. Of course, these patients may not want continuing attention, in *any* form. It should help that support groups have flourished in the United States and other countries, to the extent that any adult with a repaired cleft or other craniofacial anomaly should be able to find other adults to talk to about it. Again, a primary source for finding support groups is the Cleft Palate Foundation (1-800-24CLEFT). However, as was discussed in the section on early parent support, the onslaught of online social media has opened a new world of contact for teenagers and adults with clefts (and everybody else).

### Summary for Speech-Language Pathologists

SLPs are intricately involved in the detection of developmental problems in youngsters with clefts and are also often involved in counseling of parents both prenatally and for early parent-infant issues such as feeding. They stay "on duty" as the child progresses through infancy and toddlerhood and intervene in a timely fashion when delays in communication development are detected. This remains the case throughout childhood and the teenage years. SLPs, along with the classroom teacher, will know when behavior problems emerge. Quite often, it is the SLP who first detects either a functional problem or a previously unsuspected physical finding (e.g., submucous cleft). SLPs cannot underestimate their role in successfully launching a child with a cleft toward a happy adulthood.

It should be emphasized that, despite all the possible threats to a healthy emotional and functional life discussed earlier, most children with clefts are more similar to their peers than they are different. For the most part, the child with a cleft is just like the child next to him who does not have a cleft. If the structural

brain differences revealed in some individuals with clefts in the studies of Nopoulos et al. were consistently affecting development, we would be seeing far more language problems and behavioral manifestations in children and adolescents than we do.

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